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Salivary Duct Carcinoma of the parotid gland: A Case Report.

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Abstract :*Salivary duct carcinoma of the parotid gland is an uncommon highly aggressive tumor. About 200 cases have been reported in the English literature. Pathomorphologically, these tumors showed great similarities to ductal carcinoma of the female breast, which is why they described this tumor as "salivary duct carcinoma." We present the case of a 77-year-old patient with progressive swelling in the neck since 8 months.The MRI examination of the head showed an ill definedlesion.A malignant tumor was strongly suspected, so that a right total parotidectomy along with modified neck dissection(involving Level II-IV Lymph nodes) was performed and sent for histopathological examination.Microscopic examination concluded to a salivary duct carcinoma of the right parotid gland without lymph node involvement.*
Keywords: *Parotid gland, Salivary Duct Carcinoma*

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I. Introduction

Salivary duct carcinomas (SDC) are aggressive, high-grade salivary malignancies first described by Kleinsasser*et a*l in 1968 [1]. The tumors are characterized by a histological resemblance to ductal carcinoma of the breast. The reported incidence of SDC is 1–3% among all salivary tumors[2]. This tumor exhibits aggressive clinical behavior with a tendency for early cervical lymphadenopathies and distant metastases to the lungs and bones and thus, the prognosis of SDC is highly unfavourable[3]. Surgical resection followed by radiation is the treatment of choice, however, locoregional recurrences and distant metastases have frequently been reported [4]. The disease is rarely found in the parotid gland. The present case study reviews the clinical data of a patient with SDC in the deep lobe of the parotid gland and discusses the relevant literature. Written informed consent was obtained from the patient.

II. Case Report

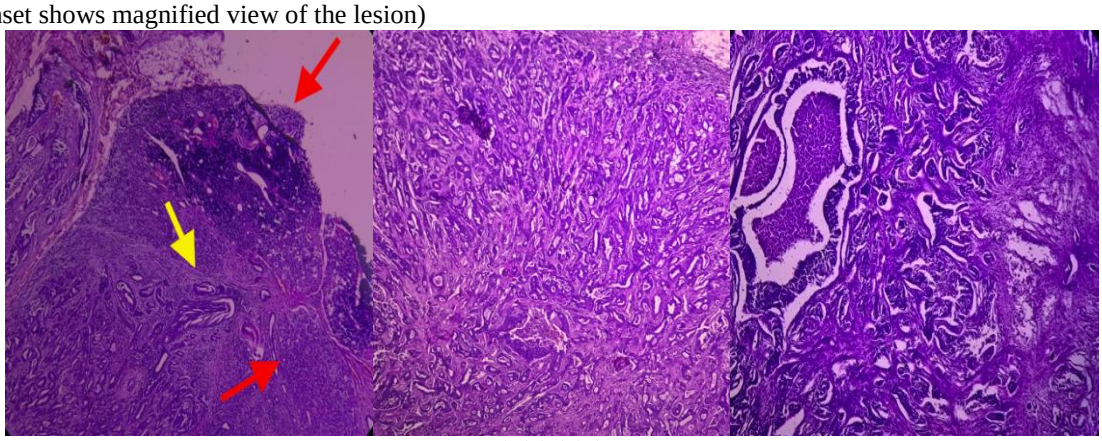
A 77-year-old male presented with a moderate, painless progressive swelling of the right neck region since 8 months that had increased in size in 10 days. The patient had no history of fever or other constitutional symptoms. A physical examination revealed a firm,non compressible but mobile lump that was not fixed to the overlying skin and was painless on palpation. The functioning of the facial nerve was within normal limits. Upon clinical examination, there was no enlarged lymph node and no abnormalities within the oral cavity. Chest X-ray was normal.Magnetic resonance imaging identified an ill defined lesion of approximately 2 cm in diameter located in the deep lobe of the parotid gland and involving the exofacial parotid gland.A malignant tumor was strongly suspected, so that a total rightparotidectomy along with modified neck dissection(involving Level II-IV Lymph nodes was performed and sent for histopathological examination.On gross examination, we received a 7x5.1x2.5 cm gland. Cut surface showed an ill-defined lesion 3x2.2x1.8cm with necrotic centre. Also received separately was an irregular grey white soft tissue piece that showed 5 lymph nodes on cutting along with blood vessels.

Microscopic examination showed invasive ductal lesions comprising pleomorphic and epithelioid tumor cells with abundant eosinophilic cytoplasm, large pleomorphic vesicular nucleus with prominent nucleoli which at places showed atypical mitotic figures,in a cribriform growth pattern. Solid and papillary areas were also noted. The surrounding stroma was dense fibrous and there was no evidence of perivascular and perineuralinvasion.Lymph node parenchyma was also observed and seems to be normal and uninvolved by malignancy. The diagnosis retained was a salivary duct carcinoma of the right parotid gland without lymph node metastasis.

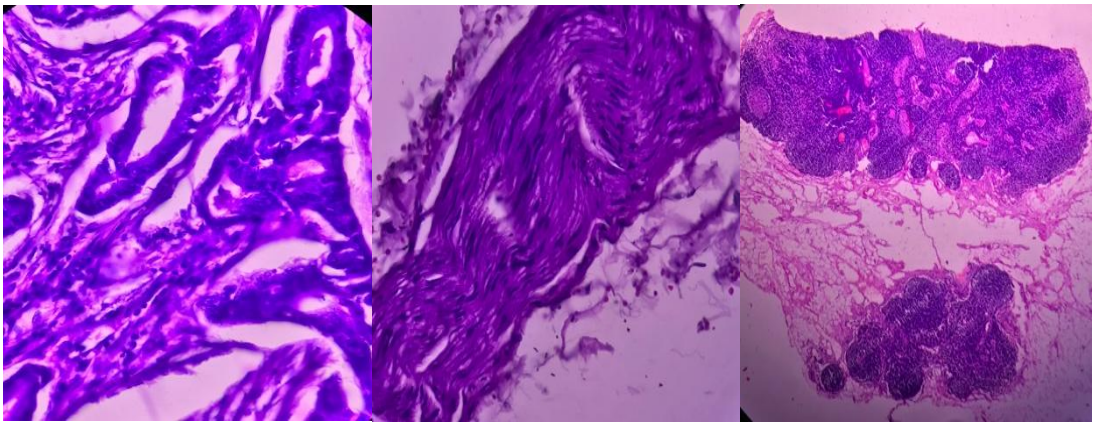
(A) (B)

Figure 1 (A: Cut surface of the parotid gland showing ill defined 3x2.2x1.8 cm lesion with necrotic centre;

B: inset shows magnified view of the lesion)



(A)(B)(C)



(D)(E)(F)

Figure 2 (A: Low power view showing foci of carcinomatous ducts(yellow arrow) invading the normal salivary gland parenchyma(red arrows); B: Predominantly cribriform pattern of malignant glands with areas of comedonecrosis(C) ; D: High power view of pleomorphic tumor cells having vesicular nuclei and prominent nucleoli; E: Uninvolved neural tissue; F: Uninvolved lymph nodes)

III. Discussion

Salivary duct carcinoma of the parotid gland is an uncommon, highly aggressive tumor. About 200 cases have been reported in the English literature. Pathomorphologically, these tumors showed great similarities to ductal carcinoma of the female breast, which is why they described this tumor as "salivary duct carcinoma."[5,6,7]. Onset is generally in the fifth or sixth decade of life, for a mean age of 60. It mainly affects men, with a male-to-female sex-ratio of 2:1. In some 20% of cases, it develops from a pre-existing benign lesion; it may, for example, result from degeneration of a pleomorphic adenoma[8,9]

Valeri*et al*[2] declared SDC to be a rare form of parotid tumor originating from the major or minor salivary glands and accounting for 0.2–2% of all salivary gland tumors. In 2005, SDC was defined as an

independent entity by the World Health Organization, labeling it as 'an aggressive adenocarcinoma, which resembled high-grade breast ductal carcinoma'. SDC was previously divided into two categories; low-grade and high-grade SDC. The low-grade SDC was recognized as a rare, cystic, proliferative carcinoma that resembled the spectrum of breast lesions, including atypical ductal hyperplasia and micropapillary and cribriform low-grade ductal carcinoma *in situ*[10]. Low-grade SDC has subsequently been defined as a classification termed low-grade cribriform cystadenocarcinoma. Under the current definition of SDC, the present case study defines high-grade SDCs as tumors that consist of solid invasive cancer nests with polygonal cancer cells surrounding a comedo-like necrosis. In the present case study, it was observed that the intraductal component of the primary foci and the malignant lymph nodes exhibited central comedo necrosis associated with a cribriform, solid or micropapillary architecture.(figure

SDC is generally a hematoxylin and eosin stain-based diagnosis, however, specific immunohistochemical and staining techniques may confirm a diagnosis in certain cases, and immunomarkers may be beneficial for future therapeutic approaches. Immunohistochemically, SDC is positive for the expression of low molecular weight CKs and epithelial membrane antigen[11].

Simpson proposed that SDCs could be classified into three main groups: Luminal androgen receptor-positive, HER2-positive and basal phenotype, which may form the basis for prognostic information and novel therapeutic possibilities[11].

The cell proliferation rate (Ki-67), the mutation of oncogenes, and the overexpression of growth factor-binding receptors like HER2/neu, p16, and p53 have been identified as important factors in the genesis of salivary gland tumors[7]. Some authors reported that Her-2/neu expression, together with an elevated proliferation rate, is associated with an unfavorable clinical course[7].Due to the infiltrative nature of SDC, radical surgery is the primary treatment; this involves the surgical removal of the tumor by parotidectomy with or without conservation of the facial nerve, followed by neck dissection to allow for ipsilateral lymph node excision. However, the rate of locoregional recurrence is high and the prognosis for survival is poor in the case of insufficient resection margins, particularly in cases with lymph node invasion[9]. Lymphatic embolism and perineural, extraparotid and/or lymphatic invasion are further indicators of a poor prognosis. Post-operative radiation therapy is mandatory in advanced cases of SDC, whereas chemoradiotherapy is generally reserved for metastatic forms of the tumor. The prognosis may be improved in tumors measuring <2 cm[9], however, the five-year recurrence-free survival rate remains at ~30%[2].

IV. Conclusion

SDC is an aggressive salivary gland malignancy for which treatment is surgical resection and neck dissection, with adjuvant radiation therapy reserved for the more advanced forms. The current report may increase knowledge with regard to SDCs. The primary clinical symptom presented by the patient in this case was a painless progressive swelling/mass in right deep parotid. Therefore, the pathological and immunohistochemical analysis of SDC is required to diagnose patients with a painless mass in the deep parotid, in order to avoid misdiagnosis. Furthermore, since SDC usually develops aggressively with the possibility of early distant metastasis and local recurrence, this indicates that surgery and postoperative radiation are beneficial for SDC patients.

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